

# Bilateral Nonsyndromic Dentigerous Cyst in a 10-Year-Old Child: A Case Report and Literature Review

## Abstract

Dentigerous cysts are the most common developmental cysts of the jaws, accounting for approximately 20% of all jaw cysts. These cysts are usually unilateral, surrounding the crowns of impacted mandibular third molars. Bilateral dentigerous cysts have been associated with syndromes such as cleidocranial dysplasia and Maroteaux–Lamy syndrome. Nonsyndromic bilateral dentigerous cysts are extremely rare, particularly in the mixed dentition. Seventeen such cases have been reported till date. This article reports the eighteenth case of bilateral dentigerous cysts involving permanent maxillary canines in a 10-year-old boy.

**Keywords:** Bilateral, deciduous, dentigerous cyst, nonsyndromic, tooth

**Bhawna Pant,  
Karla Carvalho,  
Anita Dhupar,  
Anita Spadigam**

*Department of Oral and  
Maxillofacial Pathology, Goa  
Dental College and Hospital,  
Bambolim, Goa, India*

## Introduction

Dentigerous cysts are the most common developmental odontogenic cysts, accounting for approximately 20% of all jaw cysts. It has a relative frequency of 1.44 for every 100 impacted teeth associated with cystic lesions.<sup>[1]</sup>

The concept of an inflammation-induced dentigerous cyst was initially introduced in 1996 by Benn and Altini who proposed the existence of two variants of dentigerous cysts: one developmental and the other inflammatory in origin.<sup>[2,3]</sup> The inflammatory dentigerous cyst was thought to involve deciduous teeth.<sup>[4]</sup>

The two types differ in their histologic appearance, with the uninfected developmental dentigerous cyst showing reduced enamel epithelium-like (REE) lining, whereas the inflammatory type is characterized by the presence of hyperplastic nonkeratinizing stratified squamous epithelium resembling a radicular cyst.<sup>[3]</sup> A PubMed literature search revealed that seventeen cases of nonsyndromic bilateral dentigerous cysts in the mixed dentition have been reported till date, with no established pathogenesis for the same.

## Case Report

A 10-year-old male patient presented to the Department of Oral and Maxillofacial

Pathology complaining of swelling and pain in the upper jaw for 1 month.

Extra-oral examination revealed diffuse swellings bilaterally, obliterating the naso-labial fold. The borders of the swellings were ill defined and could not be appreciated. Intra-oral examination revealed bilateral swellings on either side of the midline, obliterating the buccal vestibule. The swellings were seen extending from the mesial aspect of 52 to the distal aspect of 54 (measuring 4 cm × 3 cm) and from the mesial aspect of 63 extending to the distal aspect of 65 (measuring 4.5 cm × 3 cm), respectively. Primary teeth associated with the swellings were grossly decayed. The partial eruption of 14 and 44 was noted [Figure 1]. Swellings were tender and fluctuant on palpation. Bilateral submandibular lymph nodes were palpable, tender, and mobile. A thorough oral and systemic examination, radiographic evaluation, and pedigree analysis revealed no syndrome association.

Radiographic image analysis revealed large, well-defined bilateral unilocular radiolucencies (>1.00 cm in diameter) around the necks of unerupted permanent maxillary canines [Figure 2]. The protein estimation of the cystic aspirate was found to be 7.4 g/dl.

Based on the clinical and radiographic data, a provisional diagnosis of the bilateral

**Received:** 28 June, 2018.

**Accepted:** 07 January, 2019.

### Address for correspondence:

Dr. Bhawna Pant,  
Department of Oral and  
Maxillofacial Pathology, Goa  
Dental College and Hospital,  
Bambolim - 403 202, Goa,  
India.  
E-mail: bhawnapant001@gmail.  
com

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**Website:**  
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**DOI:**  
10.4103/ijabmr.IJABMR\_205\_18

### Quick Response Code:



**How to cite this article:** Pant B, Carvalho K, Dhupar A, Spadigam A. Bilateral nonsyndromic dentigerous cyst in a 10-year-old child: A case report and literature review. *Int J App Basic Med Res* 2019;9:58-61.

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nonsyndromic dentigerous cyst was made. The differential diagnosis included odontogenic keratocyst and unicystic ameloblastoma.

The primary teeth were extracted. Enucleation of the cysts and simultaneous extraction of the impacted permanent canines were performed.

The histopathological evaluation revealed a cystic epithelium comprising a stratified squamous nonkeratinized epithelium which was 2–3 cell layers thick and resembled the REE. Inflammation was a prominent feature in the cystic capsule. Noteworthy inflammatory changes included hyperplasia of the cystic epithelium which coincided with juxta-epithelial lymphoplasmacytic infiltrate seen in the connective tissue capsule [Figure 3].

## Discussion

Dentigerous cysts associated with permanent dentition are thought to be developmental, whereas those associated with the mixed dentition are thought to be inflammatory in origin.<sup>[4]</sup>

A histogenetic concept has implicated infected deciduous teeth in the pathogenesis of dentigerous cyst. The authors of this concept hypothesized that the peri-apical inflammation from a carious nonvital or pulpotomized deciduous tooth, in close approximation with the follicle of the permanent successor tooth can initiate the formation of a dentigerous cyst.<sup>[2,4]</sup>

This case of nonsyndromic bilateral dentigerous cyst, favors the inflammatory pathogenic pathway. This article strives to elucidate the molecular pathway initiated at the peri-apex of the bilateral infected deciduous canines and as a result differentiate the inflammatory dentigerous cyst from the developmental variant. It is justifiable to assume in this case, that as a result of long-standing apical periodontitis, a chronic inflammatory host response would be mounted within the jaw bone. The macrophages recruitment through interferon- $\gamma$  would thus result in an inflammatory cascade around the underlying follicle of the unerupted canines. The macrophage derived growth factors namely fibroblast growth factor, platelet-derived growth factor, epidermal growth factor, and transforming growth factor- $\beta$  would thus stimulate a hyperplastic response of cystic epithelium. Macrophages also release cytokines such as interleukin-1 and interleukin-6 (IL-1 and IL-6), tumor necrosis factor- $\alpha$  and  $\beta$  (TNF- $\alpha$  and TNF- $\beta$ ) which lead to bone resorption, aiding in cyst expansion. The vasoactive mediators such as prostaglandin E2, thromboxane A2 cause platelet, and mast cell degranulation. The release of serotonin, histamine along with anaphylatoxins (C3a and C5a), and neutrophil-derived radical oxygen species (OH $^-$  and O $_2^-$ ) would explain an increased colloid osmotic pressure. The resultant increase in hydrostatic pressure would thus promote the separation of REE from the crown of the tooth [Figure 4].<sup>[1,3,5]</sup>

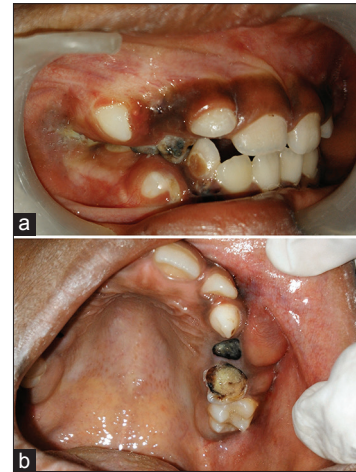


Figure 1: (a) Carious deciduous canine is seen mesial to erupting first premolar, in the right maxillary quadrant. (b) Occlusal view of the left maxillary alveolus showing buccal plate expansion and severely carious crowns of the left first and second deciduous molars



Figure 2: Orthopantomogram showing bilateral unilocular radiolucencies around the necks of unerupted permanent canines, in the maxillary jaw bone

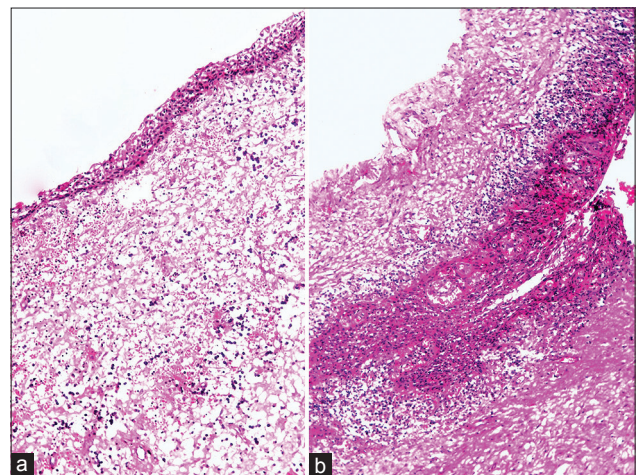
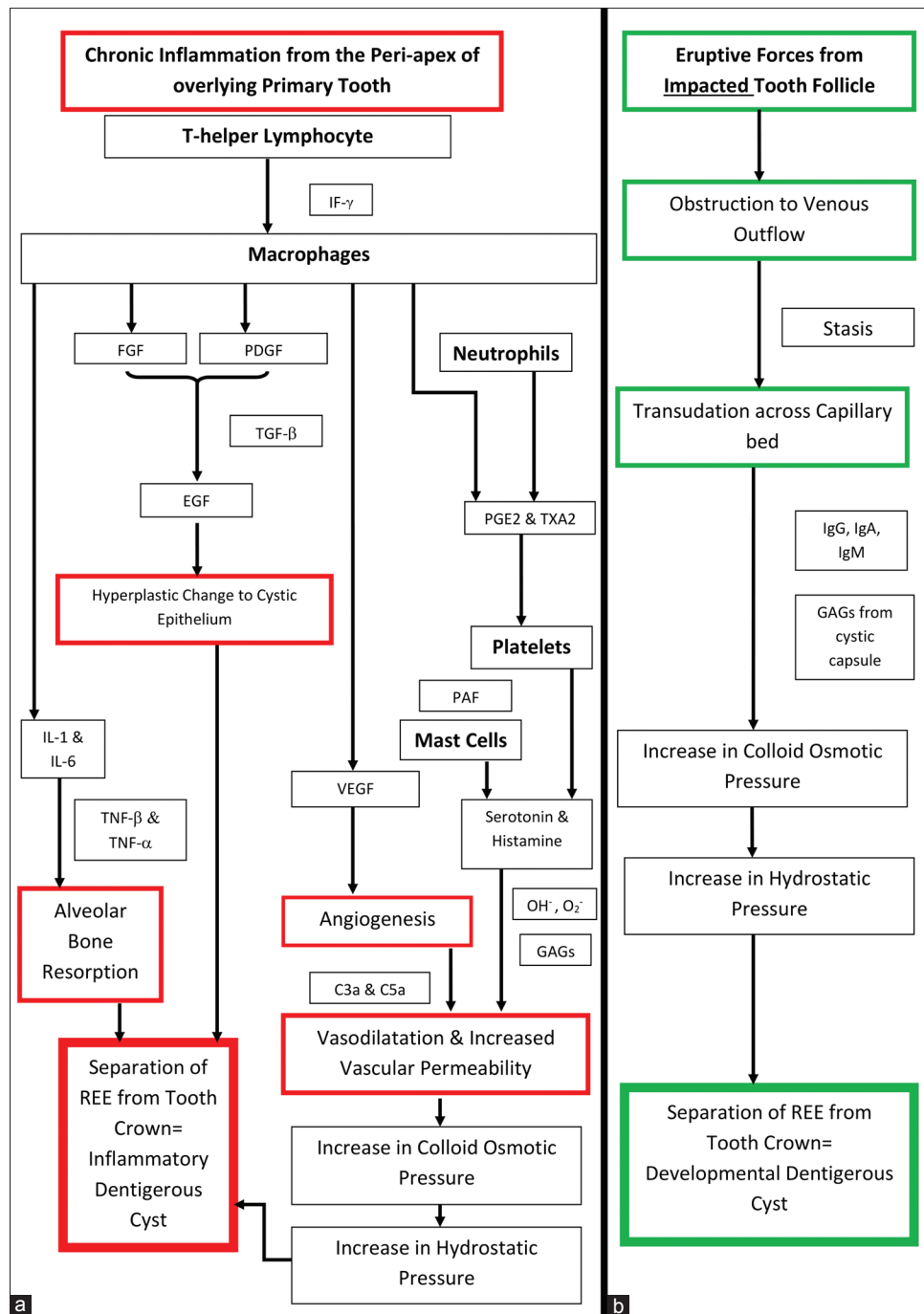


Figure 3: H and E, stained tissue section ( $\times 100$ ) (a) Cystic lining resembling reduced enamel epithelium. (b) Juxta-epithelial inflammation associated with hyperplastic cystic epithelium



**Figure 4:** Flow-chart describing the pathogenic sequence of events in dentigerous cyst formation. (a) Inflammatory dentigerous cyst. (b) Developmental dentigerous cyst

To question the presence of an eruptive force on “impacted” follicles which is the basis of a developmental dentigerous cyst in the first decade may be justified. The age of the patient in the case presented lends further support to the inflammatory origin of the dentigerous cysts. Similarly, the seventeen reported cases of nonsyndromic bilateral dentigerous cysts in the mixed dentition stage, were also seen in the first decade.<sup>[6-9]</sup>

Furthermore, some of the reported cases of nonsyndromic bilateral dentigerous cysts have shown an association

with carious nonvital and/or pulpotomized deciduous teeth. Twelve of the seventeen cases of nonsyndromic bilateral dentigerous cysts in the mixed dentition were seen involving the crown of the mandibular premolars.<sup>[6-8]</sup> The prevalence of mandible being the preferred site for a dentigerous cyst in the mixed dentition stage could be explained by the close association of the deciduous mandibular molar roots to their successor’s follicle as well as the increased susceptibility to caries seen in mandibular teeth.<sup>[4,10]</sup>

## Conclusion

The undefined pathogenesis of the dentigerous cyst seen in the mixed dentition stage should warrant further investigation. The rarity and paucity of data available in the literature for nonsyndromic bilateral dentigerous cyst make it difficult to conclude on the etiopathogenesis. However, the known differentiation of a dentigerous cyst into aggressive and malignant lesions such as ameloblastoma and mucoepidermoid carcinoma further highlights the need to research the role played by inflammation in the pathogenesis of odontogenic pathologies in children.<sup>[1,3]</sup> In addition, future studies on the role of deciduous teeth as portals of gnathic inflammation, can promote the development of novel prophylactic restorative measures in the primary and mixed dentition stages.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

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